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# TICK PARALYSIS: REGIONAL NEUROLOGICAL INVOLVEMENT CAUSED BY *HYALOMMA TRUNCATUM*

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The first record of paralytic sequelae following a tick bite appeared in 1843,[1] when Backhouse described the condition as affecting sheep in the vicinity of Sydney. It was not until 1904, however, that Malley[2] provided a definitive description of this disease, following his observations of paralysed sheep in the Eastern Province of the Cape Colony. The world-wide distribution of animal tick paralysis is now well recognized, the condition having been recorded from Australia, North America, South Africa, Macedonia, Crete, and elsewhere.

Human cases are infrequent and have only been recognized much more recently. Most accounts have come from North America and Australia. Reports have appeared of only 2 fully documented cases in South Africa, both from the Transvaal,[3,4] and in one of these reports[4] brief mention is made of personal communications concerning 2 further patients—one living in the Willowmore district of the Cape Province, and the other near Pretoria. The following case is described because it appears to be the first recorded in the Western Cape, because the offending tick was a male of the genus *Hyalomma truncatum*, and because of the development of permanent neurological sequelae.

## CASE REPORT

A Coloured youth aged 16 years, resident at Goedverwacht (near Piketberg), developed a severe pain in his right forearm, which was followed within a few minutes by inability to move his hand and fingers. He had not experienced any preceding or accompanying constitutional disturbances nor any significant trauma. On admission to Groote Schuur Hospital 10 days later (14 October 1957), his pulse rate and temperature were normal and physical abnormalities were confined to the right upper limb. Shoulder movements provoked pain in the axilla, and were voluntarily restricted. Elbow flexion and extension were moderately weakened, but the forearm flexors were grossly affected. The long flexors of the fingers were completely paralysed, and wrist flexion was barely present. The interossei and other small hand muscles were not functioning. The extensors of the wrist and fingers were only slightly weakened. Superficial cutaneous sensibility was entirely lost on the medial aspect of the forearm and hand, in the distribution of the medial cutaneous nerve of the forearm and in the cutaneous innervation of the ulnar nerve. Biceps, triceps and supinator reflexes were absent. Electrical reaction of degeneration was elicited in the following muscles: Pronator teres, flexor digitorum sublimus, flexor pollicis longus, opponens pollicis, abductor pollicis, abductor digiti minimi, dorsal interossei, and lumbricals. No alteration in motor functions could be observed to follow an intramuscular injection of 1·5 mg. of prostigmine plus 1/100 gr. of atropine.

The following results of other investigations were obtained: Sedimentation rate 12 mm. per hour (Westergren), haemoglobin 14·5 g.%, packed cell volume 51%, leucocytes 7,400 per c.mm. (polymorphs 62%, lymphocytes 34%, monocytes 3%, eosinophils 1%). The urine contained no abnormal chemical or microscopic constituents. The CSF was clear and colourless, pressure 70 mm. water, protein 10 mg.%, globulin absent, sugar normal, red cells 3 and lymphocytes 1 per c.mm. The Wassermann and Berger reactions of the blood and CSF were negative. The Weil-Felix reaction was negative. An electrocardiogram and X-ray films of the chest were normal.

In the right axilla a small hard non-engorged tick was found to be firmly attached (Fig. 1). The surrounding skin was erythematous and indurated, and some mild ecchymoses were present distal to the bite. A biopsy from the latter area showed a limited and patchy perivascular round-cell infiltration in the corium.

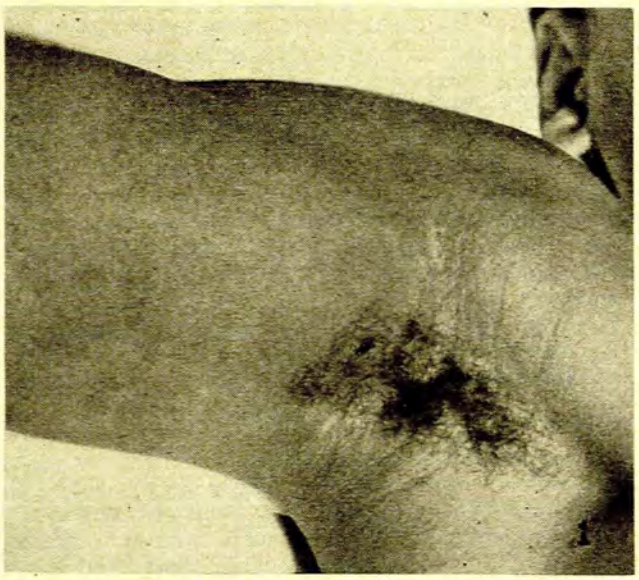

*Fig.* 1. Tick attached to right axilla. Fibres in the median and ulnar nerves were involved.

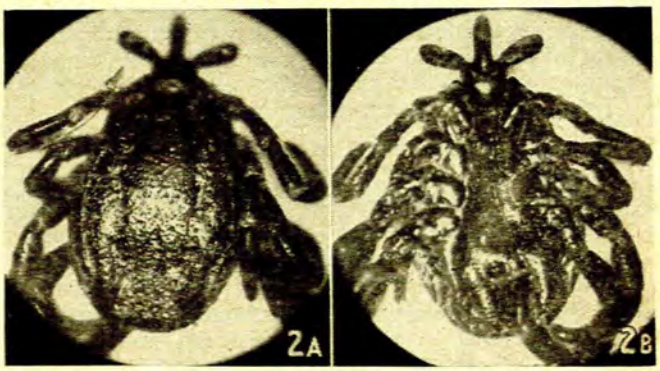


*Fig.* 2. Dorsal (A) and ventral (B) views of the tick *Hyalomma truncatum* (male).

In addition, small numbers of extravasated red cells were seen in the superficial corium, especially in relation to some of the vessels, but no evidence of necrotizing arterial lesions (Dr. M. Sacks). The tick was carefully dislodged and photographs were subsequently identified by Dr. G. Theiler as those of a male *Hyalomma truncatum* (Fig. 2).

*Subsequent course.* After removal of the tick the pain disappeared gradually during the ensuing 3 days, the area of sensory impairment contracted and sensibility was intact within 1 week. With the subsidence of local reaction, movements of the shoulder and elbow no longer caused pain, and after 1 week more were not demonstrably weak. The extensors of the wrist became virtually normal, but flexion of the wrist and fingers remained grossly weakened, and substantially unchanged since admission. Intensive physiotherapy failed to affect the situation, and when he was seen again 8 months later frank wasting had appeared in the flexors of the wrist and fingers and the small muscles of the right hand. All these muscles virtually remained completely paralysed.

## COMMENT

Tick paralysis is an important veterinary disease, but despite its prevalence little is known concerning the fundamental mechanisms of the condition. Comprehensive reviews of human involvement have appeared[5-8] and the clinical modes of presentation are well defined.

The species of tick capable of causing paralysis varies from place to place. In North America, *Dermacentor variabilis* Say, *Dermacentor andersoni* Stiles, *Amblyomma maculatum* Koch, and *Amblyomma americanum* (L) are the main offenders, in Crete *Ixodes ricinus* and *Haemaphysalis punctata*, and in Yugoslavia various Ixodid ticks.[5] In Australia *Ixodes holocyclus* Neum has been associated with cases in man as well as in animals.

In South Africa *Ixodes rubicundes* (also known as the Karoo paralysis tick) is responsible for an important paralytic illness of sheep throughout most of the Karoo, southern Orange Free State and eastern Transvaal.[9] The condition appears to be extending to previously unaffected areas. There is as yet no clear evidence that *Ixodes rubicundes* has caused paralysis in humans. The human cases hitherto described probably contracted the illness whilst residing in the Krugersdorp, Pretoria and Willowmore districts.[3,4] Ticks were identified in two of them as being respectively *Rhipicephalus simus* Koch[3] and *Hyalomma transiens*[4] (accurately classified recently and now known as *Hyalomma truncatum*.[9]) The present patient is thus the second South African case of human paralysis due to *Hyalomma truncatum*, and a point of great interest is that in both cases the ticks were males. Hyalommas are widely distributed throughout South Africa, but apart from these two cases they have not been found associated with paralysis in man or animal,[4] though they are suspected of causing both local and generalized types of paralysis in lambs in the Rift Valley of Kenya.[10]

### *Clinical Features*

It is possible that different species of ticks may elaborate distinct toxins and that this, as well as the host susceptibility, influences the symptomatology. Children are generally more susceptible and experience the illness in severe form; in Australia it has been labelled 'a dangerous disease in children'.[11] This susceptibility of the young applies also to animals. Furthermore it appears that, in sheep, the Australian tick *Ixodes holocyclus* tends to cause a severer illness than either the American *Dermacentor andersoni*, or the South African *Ixodes rubicundes*. In humans, 2 main forms of the disease may be defined, as follows:

1. The common variety, in which more or less widespread neurological involvement occurs, mainly in the form of progressive weakness with subjective but few objective sensory changes and rapid extension in Landry fashion to complete lower-motor-neurone paralysis within 48 hours. When bulbar involvement ensues, dysarthria, dysphagia and ocular palsies may proceed to respiratory paralysis and death. Sphincter functions may or may not be involved.The frequent occurrence of ataxia and of nystagmus suggests that the cerebellum or its connections may be attacked. Occasionally involvement of the encephalon is suggested by mental clouding or stupor. Significant pyrexia is rare but may occur terminally. After removal of the tick recovery is generally manifest within a few hours, and is usually complete within a week. The cerebrospinal fluid is normal, although a pleocytosis of 290 leucocytes has been described in one fatal case shortly before death,[8] and elevated CSF pressure accompanied by papilloedema and retinal haemorrhages was noted in another (non-fatal) case.[3]

2. Localized neurological involvement is a much less common variety, and only a few cases have been described.[4,11-15] Of the 7 case reports available to me, 4 patients presented with partial or total hemifacial paralysis due to tick bites in the external auditory meatus or the temporal region,[11-14] and 3 developed paralyses of arm muscles associated with tick bites in the axillae.[4,11,15] Naturally, it is rarely possible to ascertain how long a tick has been attached before paralysis ensues; in one of the patients who developed facial weakness there were good reasons for believing that the tick had entered his auditory meatus 20 days previously.[13] Apart from the two South African cases due to *Hyalomma truncatum*, only motor nerves appeared to have been involved and, in all the case reports available, complete recovery followed from 3 days to $3\frac{1}{2}$ months after removal of the tick. In Malherbe's patient[4] a small area of sensory loss persisted when the patient was last seen, $3\frac{1}{2}$ months after his discharge from hospital, but the motor weakness had resolved entirely. The case now presented is thus most unusual, if not unique, as an unequivocal example of permanent motor residua following regional tick paralysis.

### *Pathology and Pathogenesis*

From the scanty pathological data available, no characteristic pattern can be construed. Necropsies on lambs in which the disease had been experimentally induced revealed congestion of the brain and meninges, and fibrinous exudate in the ventricles.[16,17] In dogs infested with *Ixodes holocyclus*, marked congestion of anterior and posterior horns, perivascular mononuclear infiltration, and distinct neurophagia, were detected.[18] The nerve cells appeared healthy on the whole. Other authors have failed to find any histological lesions in experimental and clinical material. Pathological data on regionally paralysed patients are confined to the local lesion, and do not throw any light on the nature of the nerve involvement.

It is quite clear that female ticks of different species may transmit a substance or substances capable of causing generalized tick paralysis, but it is not at all certain what relation regional tick paralysis bears to the generalized form. In the two patients with regional paralyses where the offending ticks were identified, both were male ticks, members of the genus *Hyalomma truncatum*. In regionally involved patients reported from Australia the ticks were not available for

identification. Until Malherbe's report in 1952,[4] male ticks were thought to be inoffensive, but the two recorded examples of regional paralysis due to Hyalomma ticks suggest that the male of this genus can elaborate a toxin with a restricted local action and little or no systemic or more widespread neurological involvement. However, future cases may modify this concept.

Formerly only gravid females were considered to be capable of causing tick paralysis, and this gave rise to a widespread belief that the offending agent was initially localized within the egg, subsequently diffusing throughout the body of the tick.[19] It has however been shown that nymphal and larval forms are capable of causing paralysis.[20] Although the eggs of certain ticks, including all the Ixodae, have been found to contain a toxic substance, it is doubtful whether this is the same as the paralytic agent responsible for tick paralysis.[21] The continued presence of a tick is necessary to produce paralysis, and removal of the tick is as a rule followed by rapid recovery. This suggests continuous transmission of the toxic substance, which probably exists in high concentration in the salivary glands, and makes an infectious agent unlikely. The mode of spread to the nervous system is uncertain. Some apparent similarities between this disease and tetanus tempt one to speculate that diffusion first occurs to the regional nerves, followed by transmission (in generalized cases) along the peripheral nerves to the central nervous system.

The toxin or toxins have not been identified. Attempts at isolating an infective agent have failed, neither has an immune mechanism been demonstrated, although some immunity may occur. Paralyses induced in dogs by applying the wood tick (*Dermacentor andersoni*) have been shown to be due to failure in transmission at the neuromuscular junction. This block in transmission is probably due to failure in liberation or synthesis of acetylcholine at the nerve terminals. The paralysed muscles contract on rapid close intra-arterial injections of acetylcholine, neostigmine does not facilitate transmission in a fully paralysed muscle, and the paralysis is intensified by curare but uninfluenced by pentamethonium.[22]

*Treatment*

The only definitive treatment is immediate removal of the entire tick. The condition may persist or advance if the head is left in the skin. Other measures are essentially supportive and palliative, although claims are made for an antiserum which is used in Australia. This serum was prepared from dogs on whom *Ixodes holocyclus* had been allowed to feed and has been administered to human patients. In animals a high degree of immunity may undoubtedly occur. This may be twofold, viz. a broad non-specific immunity against ticks causing the animal to become less susceptible to tick infestation, and those ticks that do attach themselves, to engorge less avidly; and also a more specific immunity, of short duration, to the toxic agent itself.[23]

SUMMARY

A patient with regional tick paralysis of upper limb muscles is described.

Transient sensory impairment occurred, and permanent lower-motor-neurone paralysis ensued.

The offending tick was a male of the genus *Hyalomma truncatum*.

A brief account is given of the clinical and aetiological features of human tick paralysis.

I wish to thank Dr. G. Theiler for identifying the tick and for other useful information, and Prof. J. F. Brock, to whose ward the patient was admitted, and Dr. J. J. Burger, Superintendent of Groote Schuur Hospital, for permission to publish.

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